SUMMARY

A case is reported of eosinophilic granuloma involving the calvarium, buccal mucosa and inguinal skin. The diagnosis was established by histologic study of each lesion. Radiologic examinations revealed presumed involvement of the lungs and pelvis. There was no laboratory or clinical evidence of diabetes insipidus. The present case is considered another example of the disease group called Histiocytosis X, which includes those disease entities formerly called eosinophilic granuloma of the bone, Hand-Schüller-Christian disease and Letterer-Siwe disease.

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## Relief of Canker Sores on Resumption Of Cigarette Smoking

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ALTHOUGH APHTHOUS STOMATITIS—"canker sores"—usually takes the form of lesions of the oral mucosa or tongue that heal spontaneously, sometimes the lesions are multiple, confluent, recurrent and resistant to therapy. In that form the disease can torment the patient and dismay the physician who treats him.

The etiology of recurrent aphthae is obscure. Various observers have inculpated viral infection,[1,5,11] allergic sensitivity to foods,[3] hormonal influence (with exacerbation related to menses[2,4]) and trauma from weak organic acids,[8,12] but proof of any single cause is lacking.

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Numerous forms of therapy—antibiotics both locally[6] and systemically,[7] hydrocortisone,[10] a variety of topical agents and frequent smallpox vaccinations[9]—have been reported, but none with uniform success.

In these circumstances any observation, however perplexing, that might cast light on the cause or treatment of this disease merits reporting.

In the four cases herein reported, the patients had dramatic and almost complete remission of aphthae as long as they continued to smoke tobacco in cigarettes. In two cases the discovery of the phenomenon was entirely fortuitous; in the other two, in light of this observation, smoking was advised.

CASE 1. The patient, a man 58 years of age, had a history of multiple aphthae developing within a week after he stopped smoking 18 years previously. The disease followed a regular pattern: Large multiple and painful lesions would occur, sometimes ten or twelve at once, involving the tongue particularly and making eating and speaking difficult. After about two weeks they would abate, with a period of comparative comfort for three or four days, then the cycle would start again. Up until the time the series of multiple lesions began, the patient had noted a small solitary canker sore about once a month. Several months after the onset of the more severe lesions, the patient started to smoke again. Within 24 hours the pain was relieved and within three days the aphthae disappeared. Thereafter, as long as he smoked he was free of lesions except for about one small solitary ulcer once a month. Several times he stopped smoking again, and each time severe aphthae developed within a few days, then subsided when he resumed smoking. He said the phenomenon apparently was not related to the brand of cigarettes and, as a heavy smoker, he could not estimate the minimum number associated with relief.

There were no symptoms suggestive of allergic sensitivity in his history, nor could questioning elicit any specific inciting or aggravating factors related to the development of aphthae.

CASE 2. A 53-year-old man had a continuous series of large, recurrent, multiple and confluent ulcerations of the tongue and mucous membrane of the anterior part of the mouth that began within two weeks after he stopped smoking in 1954. The individual lesions took about three weeks to heal and he was never free of them. Eating and speaking were very difficult because of the pain. The patient started to smoke again in August 1956, and within two or three days the ulcers disappeared. From then on, he had occasional small single ulcers which disappeared in two or three days. He did not again stop smoking. He regularly smoked filtered cigarettes but said he believed that the brand was not a factor. He was unable to estimate the minimum number of cigarettes associated with the control of the ulcers.

There were no other symptoms suggestive of allergic disease nor were there any inciting or aggravating factors during the two years he was affected.

CASE 3. The patient, a physician 53 years of age, had had recurrent episodes of large, multiple, painful ulcerations of the tongue and oral mucosa, interfering with mastication and speech, since 1943. Individual lesions took almost two weeks to heal. He tried every suggested form of local and systemic medication, including repeated smallpox vaccinations and injections of nicotine, and even had had gold dental fillings changed to silver on the possibility that the metal played a part. No therapy was in any way effective. Although he observed that certain foods would irritate the lesions and increase the pain, the only food that he could certainly and consistently associate with production of ulceration was mushrooms. The patient also had severe allergic rhinitis and for that reason had had complete skin testing for allergens in 1947. His symptoms were perennial and the substances to which his skin reacted were mainly environmental. Results of tests with foods were equivocal, with no strongly positive response to any, including mushroom. Allergic desensitization and environmental control brought about decided improvement of the rhinitis but had no effect upon the aphthae.

In light of the experience in Case 1, the patient, who had not smoked since 1930, was advised to smoke cigarettes. Relief was immediate, dramatic and lasting. The patient found that he had no severe recurrence if he regularly smoked about five cigarettes a day, and he said that although he smoked unfiltered cigarettes as a matter of preference, he believed neither the brand nor the presence or absence of a filter affected the result. Small vesicles developed on the tongue occasionally, but never on the oral mucosa. They disappeared when he smoked more frequently. In 1957 the patient stopped smoking and the ulcerations recurred in two day. They did not subside until some ten days after he resumed smoking.

CASE 4. The patient, 38-year-old man, had been a very casual smoker between the ages of 22 and 33. During that time he had infrequent small canker sores which were never troublesome. He stopped smoking at the age of 33. Two years later severe canker sores developed. They were large and painful, always on the mucous membrane of the mouth, never on the tongue. The ulcers overlapped, and as one would heal another would develop. Speech was so difficult that the patient's work as a minister was hampered. Upon advice he began smoking cigarettes. There was immediate relief and—except for infrequent, small and painless aphthae which lasted only two or three days—no recurrence so long as he smoked regularly. He found that the ulcers recurred in three days if he stopped smoking. Then when he resumed smoking, if there was an early lesion it promptly subsided and did not recur, but a lesion that was fully developed took at least a week to subside. On questioning, the patient could not recall having had any symptoms suggestive of the allergic state, and he knew of no factors which would incite or aggravate the lesions. The relief he obtained was not associated with any particular brand of cigarettes; if he smoked four or five a day, of any brand, he remained free of symptoms.

## DISCUSSION

The association of relief of recurrent aphthous stomatitis with the smoking of cigarettes is entirely empiric. I cannot even conjecture a logical explanation of it, whether chemical, metabolic or allergic. The occurrence of occasional aphthae despite smoking suggests that the effect—if effect it be—is suppressive at best. One of the four patients had allergic disease, quite distinct from the oral ulcers, which was not influenced by smoking. Skin tests with acetone-precipitated extract of tobacco prepared in our own laboratory did not show any reaction in any case and should logically not be expected to do so. The four case reports are offered only because they seem to indicate a possible therapeutic relationship between cigarette smoking and recurrent aphthous stomatitis which further experience will support or reject.

## SUMMARY

In four cases of painful recurrent aphthous stomatitis in persons who had formerly smoked cigarettes, the disease was dramatically relieved soon after the patients began smoking again.

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